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Kuttner's tumor of the parotid gland: An unusual presentation

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ABSTRACT

Chronic sclerosing sialadenitis (CSS) (Kuttner's tumor) is a benign fibroinflammatory condition most commonly affecting the submandibular gland. It presents as hard mass that mimics salivary gland neoplasms. We report an unusual presentation of Kuttner's tumor affecting the parotid gland in order to raise the awareness of this rare and underreported entity.

1. Introduction

Chronic sclerosing sialadenitis (CSS) (Kuttner's tumor) is a chronic long-lasting inflammatory disease predominantly affecting the submandibular gland with only few papers reporting parotid presentation [1,2]. Current research considers KT an immunoglobulin G4 (IgG4)-related idiopathic lesion frequently associated with adenopathy at regional lymph nodes.

Patients present with swelling associated with meals or a completely asymptomatic hard swelling [3]. Clinically, a firm painless mass which mimics a neoplastic process or painful enlarged gland such as sialadenitis is identified [4]. Kuttner's tumor, most commonly, occurs unilaterally with indurated areas of calcification simulating malignancy [3]. Ultrasound features resemble those of a 'cirrotic liver' with diffuse involvement, multiple hypoechoic lesions against an heterogenous background and duct dilatation [5]. The CT reveals homogenous attenuation and enhancement whereas the MRI demonstrate low to moderate signal intensity [6]. The differential diagnosis includes lymphomas salivary neoplasia or acute phase of Sjogren's syndrome [6].

We present a rare case of a young female patient diagnosed with Kuttner's tumor in the parotid.

2. Case report

A 37-years old patient presented with a non-tender slow-growing mass in the left parotid present approximately 3 months without any changes in size. She denied any history of dry eyes or mouth and taste alterations. Her medical history was unremarkable.

On examination, a lump on the left tail of the parotid approximately 1×1 cm was identified with no lesions at the right side and the submandibular region. The ultrasound scan revealed a lobulated slightly ill defined hypoechoic mass measuring 2.3×1x1.6 cm (Fig. 1).

Salivary gland malignancy was suspected and a left superficial parotidectomy was conducted. Six weeks after the surgery there was a grade 2 weakness of the left facial nerve and sufficient post-operative healing.

The histology reported diffuse chronic lymphocytic sialadenitis with marked acinar atrophy and sclerosis. Reactive lymph nodes were seen adjacent to the gland. Epimyoepithelial islands and lymphotic atypia weren't seen at the specimens. Patchy periductal granulomatous inflammation was also noted. The diagnosis was of chronic sclerosing sialadenitis (Kuttner's tumor). One year after the operation she had full facial nerve function and the surgical site healed completely.

Two and a half years after the operation she developed a 1.5 cm lump in the accessory parotid when an FNA and an MRI were conducted. The FNA reported ductal epithelial cells along with small and medium size lymphoid cells and histiocytes, an appearance keeping with chronic sclerosing sialadenitis. The histology from the excision was reported as benign lobulated adipose tissue.

3. Discussion

Chronic sclerosing sialadenitis (CSS) (Kuttner's tumor) most commonly occurs in the submandibular gland with only three reported cases in the parotid gland [7–9]. In fact two of those cases involve the submandibular glands as well. In our case, the submandibular area was unaffected but a slowly growing firm swelling existed at the left parotid area. The disease is most commonly identified in the fourth to the sixth decade with a male predominance [10] whereas we present the earliest onset of the disease; a female patient at the third decade.

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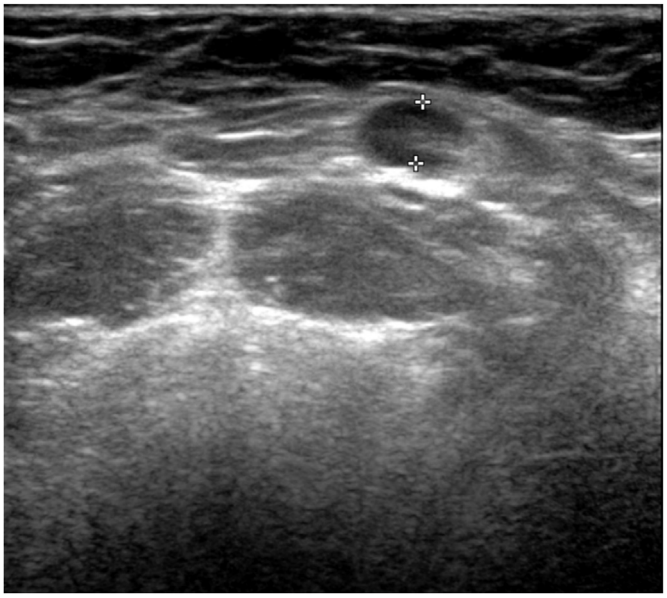

Fig. 1. The ultrasound scan of the left parotid revealed a lobulated slightly ill defined hypoechoic mass measuring 2.3 × 1 × 1.6 cm.

Table 1
Seifert's histological staging of KT.

Stage 1	Chronic inflammation and lymphocytes collection around dilated salivary ducts
Stage 2	Prominent periductal fibrosis and diffuse lymphocytic infiltration. Fibrosis centrally at the lobules and acini atrophy
Stage 3	More prominent lymphocytic infiltration with well formed lymphoid follicle, periductal sclerosis and squamous cell metaplasia.
Stage 4	Diffuse sclerosis and parenchymal loss

The exact etiology of Kuttner's tumor is not established and among the theories are salivary duct obstruction, salivary stasis, sialolithiasis, secretory dysfunctions and autoimmune response [4]. Sialoliths are found in 29%–83% of Kuttner's tumor in the submandibular gland [8, 10]. Sialoliths may cause local inflammation that contribute to the progression of chronic sclerosing sialadenitis [10]. When sialoliths cannot be found, the disease represents a primary obstructive electrolyte sialadenitis caused by secretory disorder. In our case, no sialolith was identified.

Serology and histopathology are important to differentiate from similar diseases. Organ-specific Boston consensus criteria have been reported and Kuttner's tumor is diagnosed based on the histopathological and immunohistochemistry levels. Four stages of severity have been defined based on the histopathologic examination (Table 1) [9,11]. The histopatholgical features in our case are consistent with the stage 3.

FNA for IgG4 lesions is usually nonconclusive and surgery for definitive and diagnostic purposes is recommended [10]. The operative morbidity is minimal and Kuttner's tumor doesn't tend to recur. In our case despite the complete excision a new benign lesion appeared in the accessory parotid.

4. Conclusions

Kuttner's tumor is a relatively underrecognized salivary gland enlargement in the parotid gland that should be considered in the differential diagnosis of primary parotid and facial tumors.

Ethics statement/confirmation of patient permission

Yes.

Declaration of competing interest

None.

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